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CASE REPORTS

Herpes Zoster Oticus, Ophthalmicus, and Cutaneous Disseminated: Case Report and Literature Review

Jamir Pitton Rissardo and Ana Letícia Fornari Caprara

Medicine Department, Federal University of Santa Maria, Santa Maria, Brazil

ABSTRACT

Herpes zoster (HZ) is related with the reactivation of latent varicella-zoster virus (VZV) infection. This infection is associated with HZ-oticus, HZ-ophthalmicus, and disseminated-cutaneous HZ. Here, we report a case of an adolescent male who presented with vesicular-eruptions in the left-forehead. The physical examination showed vesicles on the left V1-dermatome and external auditory canal, associated with ipsilateral periorbital oedema, peripheral VII nerve paralysis, hyperacusis, and tinnitus. Acyclovir, eye lubrication, and ophthalmic prednisolone were started. On second admission day, he developed vesicular lesions throughout the body. Polymerase chain reaction (PCR) of the draining vesicles was VZV-positive.

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Introduction

Herpes zoster (HZ) results from the reactivation of latent varicella-zoster virus (VZV) infection within a sensory ganglion. This infection might be associated with Ramsay Hunt syndrome (RHS), HZ-ophthalmicus, and disseminated-cutaneous HZ (DCHZ).[1,2] To the author's knowledge, the coexistence of these three clinical forms in an immunocompetent subject has not been reported until the present moment. Here, we report a case of an adolescent male who developed first RHS, after four days, presented HZ-ophthalmicus, and two days after, developed DCHZ.

Case report

A 16-year-old male was admitted to our hospital due to vesicular eruptions in the left forehead. The patient stated that he sought a general practitioner four days ago because he had experienced left ear pain and swelling. He was diagnosed with otitis media and was started on amoxicillin/clavulanate. In the following days, the ear pain worsened and he developed left facial weakness. One day before admission, he complained of headache, fatigue, decreased appetite, fever, and new vesicular eruptions in the left forehead.

On admission, he reported persistence of the above-mentioned symptoms and, additionally, blurred vision with one day of onset. The patient was previously healthy, and his family history was unremarkable. The physical examination showed vesicles on the distribution of the left V1 trigeminal dermatome and external auditory canal, associated with ipsilateral prominent periorbital oedema, upper and lower facial muscle paralysis, hyperacusis, and tinnitus. Laboratory tests were within normal limits. Cranial non-contrast computed tomography scan was normal, and brain magnetic resonance imaging revealed the T1-weighted left facial nerve contrast enhancement in the labyrinthine segment. Amoxicillin/clavulanate was stopped and intravenous acyclovir 2400 mg daily was started. As for his blurred vision, ophthalmology consultant observed mild punctate epithelial keratitis, and due to possible exposure keratopathy, eye lubrication and ophthalmic prednisolone acetate 1% were started.

In the second admission day, he developed erythematous papular and vesicular lesions on the face, trunk, and extremities. Blood culture was negative, and polymerase chain reaction (PCR) of the swab from the draining vesicle was positive for VZV and negative for herpes simplex virus. Acyclovir was maintained and oral cephalexin 1000 mg daily was started. From the sixth admission day onwards, the skin lesions stabilized and the patient did not have new

CONTACT Jamir Pitton Rissardo jamirrissardo@gmail.com Medicine Department, Federal University of Santa Maria, Rua Roraima, Santa Maria, Rio Grande do Sul, Brasil.

symptoms. On the 14th day, the patient was discharged home, and due to the persistent pain in the first branch of the trigeminal territory, carbamazepine 400 mg daily was started.

In the follow-up, an investigation to search an immune suppression was negative, and within three months, the residual lesions disappeared.

Discussion

James Ramsay Hunt first described the association between the geniculate ganglion VZV infection and ear sensory–motor defects. In this context, RHS is defined as a HZ-oticus infection associated with an acute peripheral facial nerve palsy, which often involves other cranial nerves (CNs).[3] The clinical manifestations include masticatory muscle weakness, taste, and sensory disturbances. However, other symptoms may occur and are thought to be related with anastomoses from the primarily affected sensory nerves and other CNs, like CN V and CN VIII, or even the spinal ganglia C2–C4.[4] The diagnosis of this syndrome is based on history and physical exam but could be supported by neurological imaging or PCR for VZV.[1,2]

HZ-ophthalmicus is other clinical form and is characterized by the involvement of the V1 division (ophthalmic) of the trigeminal nerve, where the frontal branch is most frequently affected. In about 50% of these patients, ocular involvement can occur, with epithelial keratitis and pseudodendrites that are early and transient findings.[1,5]

HZ commonly manifests as vesicular rash in dermatomal distribution. Nevertheless, it could occur also as a disseminated form defined by more than 20 skin lesions beyond the primary or adjacent dermatomes.[6] This dissemination of HZ typically occurs in hosts with cell-mediated immunity (CMI) impairment. In addition, in this group of patients, the CMI response level to VZV is the major determinant of the risk and severity of HZ.[7]

Only a few cases of RHS who developed DCHZ have been reported. We identified three cases after a thorough review of the English-language literature, and we compared these studies with the present case (Table 1).[8–10] To the author's knowledge, none of these individuals developed HZ-ophthalmicus. In this way, the present case was the first to describe a subject with RHS who developed HZ-ophthalmicus and DCHZ.

In immunocompetent hosts (ICH), the DCHZ rarely occurs, and the mechanism by which some patients are predisposed to this condition is not clearly understood. In the study of Gomez and Chernev[2], a literature review of 28 ICH with DCHZ was performed. After comparing data in Table 1, regarding only patients with RHS who developed DCHZ, with the overall data of the study of Gomez and Chernev, we observed that in our study the patients were all males and almost 15 years younger; the interval between the initial clinical symptoms until the cutaneous spread of the HZ was twice longer than the average time found by Gomez and Chernev, and postherpetic neuralgia was found more frequently in our study.[2]

The sequence of RHS, HZ-ophthalmicus, and DCHZ present in this case is rare. We propose the acronym "R.E.D. zoster" to this triad of symptoms. The letters represent Ramsay-Hunt syndrome, Eye (HZ-ophthalmicus), and Disseminated (DCHZ). We hypothesized that anastomoses from the primarily affected sensory nerves and other CNs are associated with the occurrence of this triad, since in neuropathological studies evaluating communications between CNs, the occurrence of anastomoses was a common finding within the same CN and among other CNs. Probably, our patient had anastomoses between the V and the VII CNs. More specifically and as observed in prevalence studies, the auriculotemporal branch of the V3 CN anastomoses with V1 and V2 and with the temporofacial division of the VII CN.[1,4,5,11,12]

Another interesting fact, in this case, is the onset of DCHZ after the acyclovir was started. One possible explanation could be a misleading immune-inflammatory response elicited by the intravenous infusion of the antiviral. This was already observed in other cases of HZ-ophthalmicus, and it is the rational base of the ophthalmic steroid therapy.[2,9] However, all the reported adult patients with DCHZ (Table 1) received IV acyclovir and achieved recovery. In this way, the prompt IV acyclovir administration is essential in severe cases, and patients receiving oral acyclovir should be closely monitored for the need of rapid switch to intravenous therapy in order to limit disease progression and reduce complications.

Table 1. Case reports of adult patients with RHS who developed DCHZ.

References		No.3 Nigam et al. (1980)	Yoon et al. (2013)	Chen et al. (2013)	Present case
Age(y)/sex		29/M	75/M	63/M	16/M
First clinical presentation		Skin lesions started with local redness	Left ear and C2 dermatome pain and vesicles	Left ear pain and drainage	Left ear pain and drainage
Diagnostic hypotheses and management		NR	HZ, PO antiviral agents	OME, PO amoxicillin/clavulanate	OME, PO amoxicillin/clavulanate
RHS features	Ear pain	NR	Yes	Yes	Yes
	Ear vesicles	Yes, external auditory canal	Yes, external auricle	Yes, tympanic membrane	Yes, external auditory canal
	Peripheral CN VII palsy	Yes, U	Yes, U	Yes, U	Yes, U
	CN VIII involved	No	Yes, hearing impairment, tinnitus, vertigo	Yes, dizziness on standing	Yes, hyperacusis, tinnitus
	Eyes involved	No	No	VI CN palsy, exposure keratopathy	Punctate epithelial keratitis
	Other CNs	NR	No	Laryngitis?	Left CN V1
	First dermatome affected	NR	Left CNV and C2	Left CN V	Left CN V
Other clinical manifestations		Fever	Fever	Emesis, fatigue, anorexia, night sweats, and sore throat	Headache, fatigue, anorexia, and fever
Immunocompetent or suppressed		IC	IC	IS	IC
Comorbidities		No	Diabetes mellitus, angina	Chronic lymphocytic leukaemia	No
Medications in use		No	Aspirin 100 mg/day, cilostazol 200 mg/day	Ibrutinib	No
Interval to initial clinical symptom until the CDHZ		6	11	15	6
Etiological diagnosis		Yes, positive for multinucleated giant cells	No, only clinically	Yes, punch biopsy with immunohistochemical stain positive for VZV	Yes, PCR swab from draining vesicle positive for VZV
Management, use of antiviral and/or corticoid		Yes, betamethasone	Yes, IV acyclovir 1500 mg/day for 7 days	Yes, IV acyclovir 2250 mg/day and corticosteroids	Yes, IV acyclovir 2400 mg/day for 7 days
Management specific to eye		No	No	Yes, eye lubrication and moisture goggles	Yes, eye lubrication and topical steroid drop
Outcome		Recovered	Recovered	Recovered	Recovered, but with PHN

CN: cranial nerve; DCHZ: disseminated cutaneous herpes zoster; HZ: herpes zoster; IC: immunocompromised; IS: immunosuppressed; IV: intravenous; M: male; NR: not reported; OME: otitis media with effusion; PCR: polymerase chain reaction; PHN: postherpetic neuralgia; PO: oral administration; RHS: Ramsay Hunt syndrome; U: unilateral; VZV: varicella-zoster virus; ?, aetiology not clearly defined.

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ORCID

Jamir Pitton Rissardo http://orcid.org/0000-0001-6179-2177

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